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Behavioural assessment of autism spectrum disorders in people with multiple disabilities

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Abstract

Background It is difficult to diagnose autism spectrum disorder (ASD) in people with a combination of intellectual and sensory disabilities because of overlap in behaviour. The ASD typical behaviours of people with combined intellectual and sensory disabilities are often caused by their disabilities and not by ASD. Current diagnostic tools are inadequate to differentiate between people with and without ASD when they have these combined disabilities, because tools lack norms for this population or are subjective, indirect or unable to adapt to the variety of disabilities that these people may have. Because giving a correct diagnosis is necessary for treatment and support, a new observational tool was developed to diagnose ASD in people with multiple disabilities, observation of autism in people with sensory and intellectual disabilities (OASID).

Method Observation of autism in people with sensory and intellectual disabilities was tested on 18 participants with moderate to profound intellectual disabilities, one or dual sensory impairment, with and without ASD. Two independent experts diagnosed these participants as well in order to test the psychometric properties and differentiating abilities of OASID.

Results Observation of autism in people with sensory and intellectual disabilities showed high inter-rater reliability, internal consistency of scales and content and construct validity. OASID could differentiate people with and without ASD without overlap.

Conclusions Observation of autism in people with sensory and intellectual disabilities could differentiate people with intellectual disabilities combined with sensory impairments, who clearly had or did not have signs of ASD. People with unclear signs of ADS scored in between those two groups with regard to their OASID scores. Psychometric properties of OASID are promising.

Keywords Autism, intellectual disability, multiple disabilities, sensory impairments, diagnosis, assessment

Introduction

People with sensory disabilities, intellectual disabilities (IDs) or a combination of these disabilities often show behaviours that may be symptoms of autism spectrum disorder (ASD) that in fact may be caused by their disabilities (Hobson 2005; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed 2009; Knoors & Vervloed 2011; van Gent 2012; de Vaan, Vervloed, Knoors, & Verhoeven 2013a) If it is not taken into account whether behaviours are caused by ASD or another impairment, this may lead to both underdiagnosis and overdiagnosis of ASD in this group (Jure, Rapin, & Tuchman 1991; Cass 1998; Roper, Arnold, & Monteiro 2003). The right diagnosis and proper case formulation can improve treatment,

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education and care (de Vaan *et al.* 2013a). The current study focuses on how to diagnose ASD correctly in people who have a combination of sensory and IDs (in this paper called 'multiple disabilities').

Intellectual disabilities as well as sensory impairments can both cause behaviours that are similar to ASD. There is overlap between ASD and ID (Matson & Shoemaker 2009), and this overlap becomes larger as the level of ID increases (Matson, Dempsey, LoVullo, & Wilkins 2008). This overlap is, for example, seen in stereotyped play or movements (Wing, Gould, Yeates, & Brierly 1977; Matson *et al.* 2008; Medeiros, Rojahn, Moore, & van Ingen 2014).

People with visual impairments show autistic features too (Cass 1998; Pérez-Pereira & Conti-Ramsden 1999; Hobson 2005). They may show a lack of reciprocity in social interaction (Fraiberg 1977; Dale, Tadić, & Sonksen 2014), poor use of language for social purposes and awkward pragmatic language use (Tadić, Pring, & Dale 2010), problems in understanding and using non-verbal communication (Gense & Gense 2005) and stereotyped behaviours (Tröster, Brambring, & Beelmann 1991; Warren 1994). These behaviours do not necessarily originate from ASD but are a direct result of the visual impairment.

Communication skills are affected in people with ASD but also in people with hearing impairments. Deafness leads to delays in language development, an absence of spoken language or the use of atypical language (Knoors & Vervloed 2011). Communication at home is less frequent (Vaccari & Marschark 1997), and people who are deaf can show impairments in the monitoring of conversations (Wolters, Knoors, Cillessen, & Verhoeven 2011). Delays in of theory of mind are also quite common (Peterson & Siegal 1995, 2000; Knoors & Marschark 2014). Other social impairments include deficient contact with others, disordered social imitation and impaired joint attention (Vig & Jedrysek 1999; van Gent 2012) and a preference for objects and physical attributes over social contact (Rogers & Ozonoff 2005).

People with an ID and additional visual impairments show even more problems. Among others, one finds problems in communication, social and daily living skills (Evenhuis, Sjoukes, Koot, & Kooijman 2009), such as initiating social contact or activities (Munde & Vlaskamp 2014). Maybe, the largest overlap is found in people with dual sensory disabilities (Hoevenaars-van den Boom *et al.* 2009; Dammeyer 2013). A more complete list of symptom overlap between people with ASD and people without ASD when they have sensory or IDs or a combination of both can be found in the study of de Vaan *et al.* (2013a).

Despite the presence of instruments to screen for or diagnose ASD in people with IDs (Matson & Shoemaker 2009; Matson & Williams 2014), there are still no instruments that can validly diagnose ASD in people with multiple disabilities. The available tools lack norms for people with sensory and/or IDs, and often, items are inappropriate because they require sight or hearing to pass them (Jure *et al.* 1991; Carnaby 2007; Hoevenaars-van den Boom *et al.* 2009; Matson & Shoemaker 2009).

To circumvent the aforementioned problems, Hoevenaars-van den Boom *et al.* (2009) designed a new instrument, observation of autism in deafblindness (O-ADB), for people with profound IDs and deafblindness and subsequently studied in a pilot study with 10 individuals with profound IDs and deafblindness with or without ASD. Although it differentiated ASD from no ASD successfully, only the most severe cases of deafblindness and ID were included, and administration was lengthy and rather stressful to undergo. The current study elaborated on the O-ADB, taking into account the previously mentioned limitations. The administration was made more practical by decreasing the number of tasks and thus the administration time. Lastly, the items were adapted in such a way that the people with lesser degrees of ID could also be tested. This paper describes the development of this instrument, whether it can differentiate between behaviours of people with ASD and behaviours of people without ASD and whether it can do so in a valid and reliable way.

Method

Participants

Participants were 20 clients recruited from four institutions throughout the Netherlands specialised in providing care or education to people with IDs, visual impairments, hearing impairments and deafblindness. Because of confidentiality, a contact person from every institution selected eligible clients from residential units or schools and approached their legal representatives with written information about

the study and a consent form. Staff of the institutional settings recruited participants. To warrant privacy, the exact number of people approached and reasons not to participate were not recorded. As a result, the exact response rate is unknown.

Participants qualified if the following criteria were met: (1) a moderate to profound ID (an IQ below 50), (2) a visual field of less than 20° and/or a visual acuity of less than 6/20 or complete blindness and (3) a chronological age between 5 and 55 years. Additionally, half the participants could have a hearing loss of at least 35 dB. It was requested that half of our participants were diagnosed with ASD and the other half did not. The institutional contact persons checked the institute's record for suitable participants. Participants were only considered for inclusion when assessments and diagnoses were performed by trained and licensed psychologists, psychiatrists or physicians. Because no accurate criteria existed to diagnose ASD in people with sensory and IDs, a consensus between two experts about the presence of ASD served as the gold standard. After participants were enrolled, groups were formed based on the experts' opinions whether participants had ASD or not. One of the experts was a child psychiatrist who works with deafblind children, whereas the other expert is specialised in dealing with ASD in children with IDs. Both experts made their judgements independent of each other, based on videos of the participants made for this study and a summary of the information in the participant's record. The experts also received a list of ASD typical behaviour as defined by DSM-5.

Two of the 20 participants, participants 9 and 16, were excluded from further participation because of personal circumstances. One of them had such severe motor difficulties that he was unable to perform the tasks that were expected of him. Another participant felt extremely stressed at the day of the experiment, keeping him from participating in four out of five tasks. For 9 of the remaining 18 participants (see Table 1 for an overview of participants), complete consensus regarding ASD diagnosis could be reached. For eight participants nearly reached consensus (one or both experts were uncertain about the diagnosis), for just one participant, the experts totally disagreed. So based on expert judgement, we have three groups: ASD, no ASD and doubtful.

Materials

Observation of autism in people with sensory and intellectual disabilities

For the purpose of this study, a new instrument was developed, named 'Observation of Autism in people with Sensory and Intellectual Disabilities' (OASID), to diagnose ASD in people with sensory and IDs. OASID was designed to provide a more adaptive approach in diagnosing ASD in people with multiple disabilities. OASID is a semi-structured observational instrument.

Development

After reviewing the literature and existing instruments, observations of the target population, conversations with caregivers and advice from experts in science and clinicians from the field, the differentiating characteristics for people with sensory and IDs with and without ASD were selected. These findings were compared with the diagnostic criteria from the DSM-5 (American Psychiatric Association 2013). Transformation of these criteria into testable items was carried out by reviewing the literature and items of other instruments, especially of the O-ADB (Hoevenaars-van den Boom *et al.* 2009) and the autism diagnostic observation schedule (ADOS) (Lord, Rutter, DiLavore, & Risi 1999). Preliminary versions of OASID were reviewed by all authors and authors of the O-ADB and were discussed with caregivers and other researchers, before the final version was administered to participants.

The first version OASID differentiated between participants with and participants without ASD. However, inter-rater reliability was too low, and internal consistency for one sub-scale was insufficient (de Vaan, Vervloed, Knoors, & Verhoeven 2013b). Subsequently, OASID item descriptions and the divisions of items across scales were carefully and critically reviewed and revised, aiming for an inter-rater reliability that was at least substantial (Cohen's kappa = 0.6) in the typology of Landis and Koch (1977) and an internal consistency of at least 0.7 (Nunnally & Bernstein 1994). These revisions resulted in the current version.

Procedure of administration

Participants are invited with a familiar caregiver to an assessment room, where the researcher awaits them

Table 1 Participants (excluding participants 9 and 16)

Number	Age (years)	Sex	Intellectual disability	Visual acuity (in the better eye or both eyes)	Hearing loss (in the better ear or both ears)	Syndromes or relevant medical conditions
1	11	Male	Profound	13/20	Profound hearing loss	Disabilities caused by meningitis 4 days after birth
2	24	Female	Severe	4/10	>110 dB	Congenital rubella syndrome
3	56	Female	Moderate	Blindness	90 dB	Congenital rubella syndrome
4	55	Male	Severe	3/100	105 dB	Cerebral visual impairment
5	27	Female	Profound	3/100	85 dB	Retinopathy of prematurity
6	26	Female	Severe	Severe visual impairment	112 dB	Congenital rubella syndrome
7	39	Male	Severe	1/10	Sensitive to sudden and loud noises	—
8	38	Male	Severe	Blindness, no light perception	No hearing loss	—
10	31	Female	Severe	3/20	No hearing loss	—
11	19	Female	Profound	8/100	No hearing loss	De Morsier syndrome
12	17	Female	Severe	4/10	Reactions to >45 dB	Wolf–Hirschhorn syndrome
13	18	Male	Profound	3/10	30 dB	Down syndrome
14	25	Female	Severe	3/20	No hearing loss	Near sudden death syndrome
15	19	Female	Severe	Blindness, no light perception	No hearing loss	—
17	29	Female	Moderate	Blindness, no light perception	Moderate hearing loss	—
18	23	Female	Moderate	Moderate visual impairment	Deafness	—
19	32	Male	Severe	Blindness, mild light perception	81 dB	Congenital rubella syndrome
20	50	Male	Moderate	2/10	Reactions to >108 dB	Marshall–Stickler syndrome

with several tasks. While the researcher is administrating the tasks, the familiar caregiver is present the whole time for comfort and to assist the researcher if necessary. This was carried out to prevent stress, discomfort and communication problems. Administrating the tasks lasted between 25 min and 1 hour, and this was recorded on video. No scoring occurred during administration of the tasks. Video recordings were scored and observed afterwards.

The tasks

Observation of autism in people with sensory and intellectual disabilities consists of five tasks of which an example of which can be found in Table 2. Tasks were partly inspired by the O-ADB (Hoevenaars-van den Boom *et al.* 2009) and the ADOS (Lord *et al.* 1999). All tasks were intended to elicit specific behaviours, using play materials and toys, that are expected to differentiate between people with and without ASD. All items were based on literature and clinical experience with children with and without ASD and multiple disabilities. An example is given in Table 2. The item described in Table 2 is based on the ASD criteria that people with ASD often show severe preoccupations and have difficulties in breaking routines or small changes (Turner 1999; American Psychiatric Association 2013). This is why a familiar play object was brought to the test to be later on removed to further continue playing with other objects. In people with ASD, this object could evoke preoccupied playing or no playing at all (because it is not part of their routine at that time), and removal of the object could induce stress (American Psychiatric Association 2013). The OASID scoring options take these behaviours into account.

Table 2 Example of observation of autism in people with sensory and intellectual disabilities task and corresponding question and answering possibilities

Task	Example question	Behaviour resulting in score of 0	Behaviour resulting in score of 1	Behaviour resulting in score of 2
The participant has played with their own toy or object, and after a while, the experimenter took away the object.	How does the person respond to the removal of their own object?	The person does not show anger, sadness or frustration as a response to the removal of their object.	The person shows some anger, sadness or frustration as a response to the removal of the object, or briefly clings to it, but gives the object away within three attempts of the experimenter.	The person shows anger, sadness or frustration as a response to the removal of their object, and he or she may cling to the object and refuses to give the object away even after three attempts of the experimenter.

Within one task of OASID, several ASD typical behaviours can be elicited. As a result, only a few tasks with a limited set of materials were included in OASID, avoiding overstimulation of participants. A distinguishing feature of OASID is the adaptive approach that is used in the administration. Administration of the tasks is adjusted to the abilities, severity of intellectual and sensory disabilities and communication style of individual participants. For example, different prompts are used to elicit joint attention, such as eye gaze and physical direction towards an object. The difficulty level of some test items can also be adapted. For participants with a moderate ID and a slight visual impairment, a difficult puzzle can be given, while for people with profound IDs and blindness or severe motoric difficulties, the task is simplified to putting puzzle pieces in a bucket. The presentation of the puzzle pieces is also adjusted to the participant's abilities. For instance, someone who is blind is presented with the pieces by giving them in their hands or bringing their hands to the pieces, while sighted persons are shown the pieces. Because the objective of the task was to judge the interaction of the person with the experimenter instead of solving the puzzle that was not the objective of the task, but to see how the person interacts with the researcher or asks for help, these adjustments in difficulty could be made. Mode of communication is adjusted to each individual's possibilities. Verbal participants were spoken to, while for others, simple hand gestures and signs will be used, and for other participants, communication consists of handing over objects. Finally, an important principle in administrating OASID is that the researcher did not only attempt to elicit behaviour in the participants but also wait for initiatives of the participant for social contact and responds to those. This adaptive approach made administration suitable for a broad range of participants.

The scoring form

After administration of the tasks, recordings of the session were used to score ASD typical behaviours. Of the 40 items, 29 items scored ASD typical behaviours seen in the five tasks. The remaining 11 items were holistic items scoring behaviours that occurred during the entire administration.

The 40 items were scored on a 3-point Likert scale (0, 1, 2) where a higher score indicated more ASD typical behaviour. Elaborate descriptions for each answering possibility were provided. An example question can be found in Table 2. A final score is obtained by adding all item scores and by calculating two scales and seven sub-scales that are in line with DSM-5 criteria; see Table 3.

Other measures

In addition to OASID, two questionnaires were filled out by parents or caregivers to study convergent and divergent validity. The Autisme-en verwachte stoornissenschaal-Z-revisie (AVZ-R) is the Dutch version of the Pervasive Developmental Disorder in Mental Retardation Scale that is a short questionnaire often used to diagnose ASD in children with IDs (Kraijer & Bildt 2005). This scale does not take into account possible sensory impairments. Also, parents or caregivers filled out the Dutch translation of the list

Table 3 Scales and sub-scales of observation of autism in people with sensory and intellectual disabilities along DSM-5 criteria

Scale	Number of items
A. Social behaviour and communication	21
1. Reciprocity	9
2. Communication	3
3. Relationships	9
B. Repetitive and stereotyped behaviour	19
1. Stereotyped behaviour	7
2. Insistence on sameness	6
3. Restricted interests	3
4. Reactivity to sensory input	3

of behavioural signs of disturbed attachment in young children (Stor & Storsbergen 2006), originally described by Boris and Zeanah (2005). This list was chosen because behaviours of disturbed attachment may appear similar to ASD typical behaviour but are not equal and differentiation between the two should be made (Rutgers, Bakermans-Kranenburg, van Ijzendoorn, & van Berckelaer-Onnes 2004; Zeanah, Smyke, Koga, Carlson, & The Bucharest Early Intervention Project Core 2005).

Procedure

This study was approved by the region's medical ethical review board and legal representatives, or parents signed the informed consent form prior to participation. After this, OASID was administered in a room without distractions, other clients and familiar toys and recorded on video as described previously. The caregiver that was present did not interact with the participant unless necessary and filled in the other questionnaires, while the experimenter performed the tasks with the participant. The recorded sessions were independently scored afterwards by the first author and two trained raters, all of them naive to the participant's background and possible ASD diagnosis.

Statistical analyses

Because of the ordinal nature of the scoring system, non-parametric tests were used for analyses. For reliability and validity measures, all 18 participants were included. In case of multiple comparisons, a significance level of .01 was used to avoid chance capitalization.

Item distributions for OASID were checked to make sure that all items had discriminating power. Items with identical scores for 90% or more of the participants were excluded. Only for two items, 87% of participants received the same score, and all of the other items had a lower percentage of identical scores. Therefore, no items were excluded.

For the inter-rater reliability, Cohen's kappa (Cohen 1960), the intraclass correlation coefficient and weighted kappa (Fleiss & Cohen 1973) were assessed. Because OASID scores are ordinal, a two-point difference between raters is worse than a one-point difference, and weighted kappa takes this into account (Fleiss & Cohen 1973). Cronbach's alpha was not calculated for individual sub-scales but only for the total scale and the scales 'Social behaviour and communication' and 'Repetitive and stereotyped behaviour', as the small number of items on each sub-scale might negatively influence Cronbach's alpha and underestimate reliability (Cortina 1993).

Results

Reliability

Inter-rater reliability

Videos of all participants were rated by the first author, and a random selection of seven videos was rated as well by two undergraduate students in pedagogical and educational sciences after they had received training in scoring OASID. First, between student raters, percentage of agreement was 77.8%, the corresponding Cohen's kappa was .65 and weighted kappa was .75. The intraclass correlation coefficient, taking into account scores of all three raters, was .69. All values indicated a substantial to good inter-rater reliability (Altman 1991; Landis & Koch 1977).

Internal consistency

Table 3 shows that a priori OASID consists of two scales and seven sub-scales. To measure the internal consistency of items on these scales, Cronbach's alpha was calculated. Cronbach's alpha for OASID completely was .94, for the first scale 'social communication and interaction', it was .94 and for the second scale 'repetitive and stereotyped

behaviour', it was .79. These values indicated good to excellent internal consistency of items (Kline 1993; Nunnally & Bernstein 1994).

Validity

Content validity

A high content validity was pursued by ascertaining that all testable characteristics of ASD were included in OASID. This was carried out in multiple steps. First, a theoretical framework was built based on recent literature about characteristics of ASD in people with sensory and IDs. Second, existing instruments to diagnose ASD such as the O-ADB (Hoevenaars-van den Boom *et al.* 2009) and the ADOS (Lord *et al.* 1999) were studied to transform the autistic characteristics into testable items. Third, these items and tasks were discussed with experts and clinicians from the field of multiple disabilities and adjusted according to their advice. Finally, we compared our items with criteria in DSM-5 (American Psychiatric Association 2013), resulting in scales and sub-scales that correspond to these criteria. All DSM-5 criteria for ASD were included. Preliminary versions of OASID were reviewed by all authors and the authors of the O-ADB and discussed with caretakers and clinicians. The two experts who assessed the participants for ASD were not involved in the development of OASID.

Construct validity

Observation of autism in people with sensory and intellectual disabilities was compared with the AVZ-R for convergent validity and with the list of behavioural signs of disturbed attachment for divergent validity. There was no significant correlation between OASID scores and the list of disturbed attachment, $r = .46$ ($P = .57$). Although disturbed attachment behaviour may appear similar to ASD symptoms on the surface, disturbed attachment behaviours are not equal to ASD signs, so this lack of correlation indicated good divergent validity.

The AVZ-R is designed for people with IDs alone. It also measures ASD symptoms, and a significant positive correlation was expected; therefore, one-tailed correlation was calculated. The correlation between OASID and the AVZ-R was significant and moderately strong, $r = .40$ ($P = .049$), indicating a moderate divergent validity.

Differentiation

In this study, the gold standard for ASD was based on consensus of expert judgements. Of the 18 participants, the experts reached consensus in nine participants. For the remaining nine participants, experts either disagreed (one scored yes and one scored no), both doubted the diagnoses or one of the experts doubted, while the other was certain (Fig. 1).

For Fig. 1, five groups were made, with increasing certainty regarding ASD diagnosis. Group 1 contains all the participants without ASD, according to both experts, followed by group 2 where one expert doubted and the other was certain of no ASD. Participants in group 3 consist of participants where both experts doubted or they disagreed. In group 4, one expert doubted, and the other was certain that they had ASD. Finally, participants in group 5 were people with ASD (according to both experts). Two horizontal lines were drawn, corresponding to the lowest score of someone with ASD and the highest score of someone without ASD (groups 5 and 1). As can be seen, there is a large gap between people with

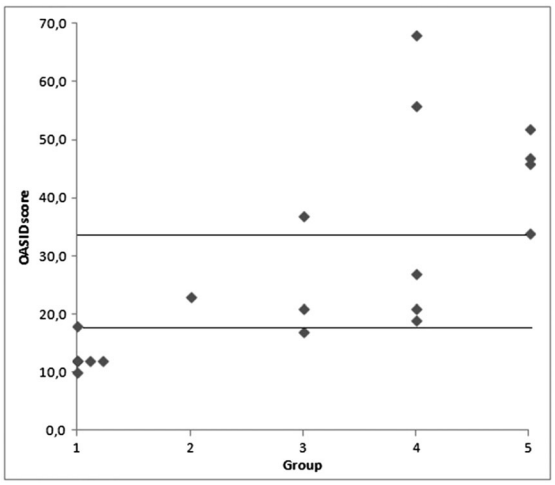

Figure 1 Groups are (1) consensus by two experts of no ASD, (2) no ASD according to one expert, the other doubts, (3) both experts doubt ASD or one was certain of ASD and the other was certain of no ASD, (4) ASD according to one expert and the other doubts, (5) consensus by two experts of ASD.

and without ASD, and most participants with doubt regarding their diagnosis (groups 2–4) scored in-between these lines. The differentiation between ASD and no ASD is further confirmed with the results from non-parametric tests; see Table 4, which showed significant differences between the ASD and no ASD group for total score, both scales and three of the sub-scales as well.

Of the nine participants that could not receive a definitive diagnosis of ASD or no ASD, five scored between the two horizontal lines. This is not surprising; as experts doubt someone's diagnosis, OASID scores are less conclusive as well. However, there are four cases that caused doubt among experts but scored below the lower or above the upper horizontal line, meaning that despite expert doubt, OASID could diagnose them. One expert doubted the ASD diagnoses of the two participants that received the highest OASID score of all participants. The other expert was certain that they had ASD. It appeared that the expert in deafblindness, a psychiatrist, was the expert who had some doubts because he believed that their impairments were not caused by ASD but by their ID. The other expert, who has expertise in ASD in people with IDs, however, was certain that they had ASD. The other two participants that score outside the horizontal lines only did so with a few points that may be coincidental.

Table 4 Differences on observation of autism in people with sensory and intellectual disabilities (OASID) between people with and without autism spectrum disorder (ASD)

Scale	ASD *M*	no ASD *M*	*P*
A. Social behaviour and communication	28.3	6.2	.008*
1. Reciprocity	10.8	1.2	.008*
2. Communication	4.8	1.4	.008*
3. Relationships	12.8	3.6	.008*
B. Repetitive and stereotyped behaviour	16.5	6.6	.008*
1. Stereotyped behaviour	7.3	1.2	.008*
2. Insistence on sameness	6.0	4.0	.143
3. Restricted interests	1.8	0.6	.365
4. Reactivity to sensory input	1.5	0.8	.143
OASID total score	44.7	12.8	.008*

Diagnosis ASD given by expert consensus.
*Significance at .01 level, one tailed.

These cases were the two participants that the experts disagreed on, one expert was certain of ASD and the other was certain that there was no ASD.

Discussion

Currently, no valid instruments are available to diagnose ASD in people with a combination of sensory and IDs in a wide range. Diagnosing people correctly is important and necessary to contribute to people's needs in living conditions, support and treatment. Because of a large amount of variability in communicative abilities and severity of disabilities, a more adaptive diagnostic approach is needed to adapt to the heterogeneous skills and abilities of people with multiple disabilities. For example, sharing of attention is often carried out in a visual manner, whereas a tactile approach would be more suitable for some people with multiple disabilities (Neerinckx, Vos, Van Den Noortgate, & Maes 2014), and thus, an individual approach is necessary. OASID was designed in this way, and the preliminary findings indicate that it can successfully differentiate between people with multiple disabilities with and without ASD. Not only was the average total score of participants with ASD significantly different from participants without ASD, there were no overlapping scores between these groups.

Consensus among two experts was used as a gold standard for ASD diagnoses. In the cases where an expert doubted whether the participants had ASD, OASID scores pointed in either direction. In one case, doubt seemed to be related to the experts' background. To make sure of giving the right label, only if both experts agreed, participants were included in analysing differences between groups. A limitation of this method is that the two experts used OASID video material, in addition to information from the participant's records, to base their decisions on. Because the OASID raters and the experts watched the same videos, their judgements were not completely independent. However, the experts did not see the OASID questionnaire and scoring rules, making contamination unlikely. At the moment, this is the most appropriate method for diagnosis, as only participants for whom complete consensus was reached were taken into account and no valid diagnostic instruments existed yet for this group.

The total score on OASID, as well as both the Social Behaviour and Communication Scale and the

Repetitive and Stereotyped Behaviour Scale, can differentiate people with and without ASD, when the diagnosis is based on expert consensus. On the first scale, all sub-scales do too. Three out of the four sub-scales in the Repetitive and Stereotyped Behaviour Scale showed no significant differences, but the total for this scale did.

Especially, the sub-scales on the social communicative domain showed large differences for people with these disabilities, which is consistent with the earlier research findings with the O-ADB (Hoevenaars-van den Boom *et al.* 2009). The lack of differences on sub-scales of repetitive and stereotyped behaviour may be caused by the fact that in people with sensory disabilities, IDs or a combination of these disabilities, these factors are quite common aspects of their behaviour (e.g. Andrews & Wyver 2005; Fraiberg 1977; Jan, Freeman, & Scott 1977; Murdoch 1997) and may therefore not be differentiating factors in this population, as opposed to in people with IDs alone (Matson *et al.* 2008). Nevertheless, the total score is differentiating for the two groups; perhaps, if a larger sample had been included, the differences on these sub-scales could have been larger as well. Additionally, some of the sub-scales consist of only three items. Perhaps, clear differences between people with and without ASD on these small sub-scales are not necessary, and a diagnosis can better be based on the total score. The sub-scales may potentially be used for individual treatment purposes, for it can easily be seen in which area the person is impaired the most and needs additional support. After all, the most important goal is to help everyone to obtain the treatment and support that are optimal for each individual.

Two of the 20 participants were unable to finish OASID, and for nine others, the diagnosis was unclear, but after applying our tentative thresholds, this number was reduced to five. These five participants would receive a diagnosis of mild ASD vs. severe ASD in the people above the threshold. With the help of OASID, the number of ASD signs can be placed on a continuum from no via mild to severe ASD. Probably, this provides a more appropriate way of categorising ASD symptoms as opposed to dichotomising in ASD or no ASD, for this spectrum is also found in DSM-5 (American Psychiatric Association 2013). Despite two outliers, OASID scores were consistent with expert consensus. All participants that received an expert consensus of ASD or no ASD received the same diagnosis with OASID. Future studies should bring down the number of persons for whom OASID and experts disagree.

The lack of diagnostic means for people with multiple disabilities to compare OASID with was solved by using expert consensus as a gold standard for the ASD diagnosis. However, in many cases, only one of the experts was unsure, while the other was confident of his or her diagnosis. This is not surprising as the current target population is difficult to diagnose, because of the aforementioned overlap in behaviour between people with and without ASD (e.g. Cass 1998; Hoevenaars-van den Boom *et al.* 2009; Vervloed & Knoors 2011; de Vaan *et al.* 2013a). Additionally, the fact that ASD occurs on a spectrum (American Psychiatric Association 2013) and is not a dichotomous label can explain the cases in which experts were uncertain about several of the participants. Because of these difficulties and that they did not consult each other, reaching consensus in half of the participants is an asset and confirms reliability of their diagnoses.

The fact that OASID can differentiate people with and without ASD would be meaningless without evidence of good reliability and validity. Despite the small sample and the fact that this measure is only semi-structured, evidence for substantial to good reliability was found. Unfortunately, content validity could not be supported by statistical tests because of the small sample size, but other steps were taken to assume good content validity. The measures used for construct validity preliminary showed that OASID is a valid tool, as both convergent and divergent validity were indicated. A low correlation was found between OASID and the Pervasive Developmental Disorder in Mental Retardation Scale (Kraijer & Bildt 2005), which was expected as both instruments aim to measure ASD, yet in a different group. Divergent validity was confirmed by finding no correlation with the list of disturbed attachment behaviours (Boris & Zeanah 2005). This was also expected because despite ASD typical behaviours in people without a secure attachment (Rutgers *et al.* 2004; Zeanah *et al.* 2005), people with ASD can still show signs of a secure attachment (Rutgers *et al.* 2004), so a correlation had to be absent to indicate divergent validity. Inter-rater reliability was substantial, which is especially promising considering that the observations were carried out by

two undergraduate students with a minimum amount of experience with ASD and the target population.

A limitation of the current study is the small sample size of 18 participants. Given that OASID is in the early stages of development and the potential target group is small altogether, the low number is acceptable, because we had to reserve potential participants for future research with OASID. All participants were recruited from institutional settings or schools, potentially harming representativeness of the sample. In the Netherlands, however, people with these types and combinations of disabilities primarily live in institutional settings (Evenhuis, Theunissen, Denkers, Verschuure, & Kemme 2001), and as such, the sample is representative of the study population. Representativeness was further enhanced by recruiting participants from different settings throughout the country.

The O-ADB was a successful tool in differentiating people with and without autism when participants were deafblind and had profound IDs (Hoevenaars-van den Boom *et al.* 2009). Our goal of broadening the target population to people with less severe intellectual and sensory disabilities was reached by including also people without auditory impairments and with a moderate and severe ID. OASID consisted of a fewer number of tasks than the O-ADB, making the administration also less tiresome and stressful for participants.

Benefits of OASID as opposed to other diagnostic tools are that it is specifically designed for this complex group of multiply disabled persons and that it uses observations of a situation in which ASD typical behaviours are evoked from the participants as opposed to observations performed in natural surroundings or to questionnaires. The benefit of an observational tool as opposed to a questionnaire is that actual behaviour of a participant is tested vs. the interpretations of the behaviour by only one rater. Where testing in a natural setting complicates the potential recognition of ASD typical behaviour, this study showed that testing in an experimental setting with OASID makes it possible to recognise ASD. Additionally, existing questionnaires may not be adaptive enough to account for the variability often found in this population of multiply disabled people, while administration of OASID is adjusted to the individual participant. Another advantage of OASID is that its scales are consistent with DSM-5 criteria (American Psychiatric Association 2013), and its scores reflect the continuum or spectrum that DSM-5 also proposes. Regardless of these positive results, it remains important that OASID is integrated in a broader diagnostic assessment, including multiple tests and using more than one informant (Carnaby 2007).

It must be noted that this study was only the first step in the development of OASID and more research is necessary. Our results show that in a small group of participants, OASID could differentiate between people with and without ASD when they have IDs and sensory impairments and potentially reduce the group of people for whom there are doubts.

References

Altman D. G. (1991) *Practical Statistics for Medical Research*. Chapman & Hall, London, UK.

American Psychiatric Association. (2013) *Diagnostic and Statistic Manual of Mental Disorders, DSM-5*, 5th edn. American Psychiatric Publishing, Washington, DC.

Andrews R. & Wyver S. (2005) Autistic tendencies: are there different pathways for blindness and autism spectrum disorder? *British Journal of Visual Impairment* **23**, 52–57. doi: 10.1177/0264619605054776

Boris N. W. & Zeanah, C. H. (2005) Practice parameter for the assessment and treatment of children and adolescents with reactive attachment disorder of infancy and early childhood. Retrieved from www.aacap.org

Carnaby S. (2007) Developing good practice in the clinical assessment of people with profound intellectual disabilities and multiple impairment. *Journal of Policy and Practice in Intellectual Disabilities* **4**, 88–96. doi: 10.1111/j.1741-1130.2007.00105.x

Cass H. (1998) Visual impairment and autism. *Autism* **2**, 117-38. doi: 10.1177/1362361398022002

Cohen J. A. (1960) A coefficient of agreement for nominal scales. *Educational and Psychological Measurement* **20**, 37–46.

Cortina J. M. (1993) What is coefficient alpha? An examination of theory and applications. [Article]. *Journal of Applied Psychology* **78**, 98–104.

Dale N. J., Tadić V. & Sonksen P. (2014) Social communicative variation in 1–3-year-olds with severe visual impairment. *Child: Care, Health and Development* **40**, 158–64. doi: 10.1111/cch.12065

Dammeyer J. (2013) Symptoms of autism among children with congenital deafblindness. *Journal of Autism and Developmental Disorders* 1–8. doi: 10.1007/s10803-013-1967-8

de Vaan G., Vervloed M. P. J., Knoors H. & Verhoeven L. (2013a) Autism spectrum disorders in people with sensory

and intellectual disabilities symptom overlap and differentiating characteristics. Fitzgerald M. (Ed.) In, *Recent Advances in Autism Spectrum Disorders – Volume I*. InTech, Retrieved from http://www.intechopen.com/books/recent-advances-in-autism-spectrum-disorders-volume-i/autism-spectrum-disorders-in-people-with-sensory-and-intellectual-disabilities-symptom-overlap-and-d.

de Vaan G., Vervloed M. P. J., Knoors H. & Verhoeven L. (2013b) Differentiating people with and without autism when they have intellectual and sensory disabilities. Paper presented at the American Association on Intellectual and Developmental Disabilities (AAIDD) 137th annual conference: Race to catch the future, Pittsburgh, PA.

Evenhuis H. M., Sjoukes L., Koot H. M. & Kooijman A. C. (2009) Does visual impairment lead to additional disability in adults with intellectual disabilities? *Journal of Intellectual Disability Research* **53**, 19–28. doi: 10.1111/j.1365-2788.2008.01114.x

Evenhuis H. M., Theunissen M., Denkers I., Verschuure H. & Kemme H. (2001) Prevalence of visual and hearing impairment in a Dutch institutionalized population with intellectual disability. *Journal of Intellectual Disability Research* **45**, 457–64. doi: 10.1046/j.1365-2788.2001.00350.x

Fleiss J. L. & Cohen J. (1973) The equivalence of weighted kappa and the intraclass correlation coefficient as measures of reliability. *Educational and Psychological Measurement* **33**, 613–9.

Fraiberg S. (1977) *Insights from the Blind*. Basic Books, New York, NY.

Gense M. H. & Gense D. J. (2005) *Autism Spectrum Disorders and Visual Impairment: Meeting Student's Learning Needs*.American Foundation for the Blind Press, New York, NY.

Hobson R. P. (2005) Why connect? On the relation between autism and blindness. In *Autism and Blindness: Research and Reflections* (ed. L. Pring), pp. 10–25. Whurr Publishers, London, United Kingdom.

Hoevenaars-van den Boom M. A. A., Antonissen A. C. F. M., Knoors H. & Vervloed M. P. J. (2009) Differentiating characteristics of deafblindness and autism in people with congenital deafblindness and profound intellectual disability. *Journal of Intellectual Disability Research* **53**, 548–58. doi: 10.1111/j.1365-2788.2009.01175.x

Jan J. E., Freeman R. D. & Scott E. P. (1977) *Visual Impairment in Children and Adolescents*. Grune & Stratton, New York, NY.

Jure R., Rapin I. & Tuchman R. F. (1991) Hearing-impaired autistic children. *Developmental Medicine & Child Neurology* **33**, 1062–72. doi: 10.1111/j.1469-8749.1991.tb14828.x

Kline P. (1993). *The Handbook of Psychological Testing*. Routledge, London, UK.

Knoors H. & Marschark M. (2014) *Teaching Deaf Learners, Psychological and Developmental Foundations*. Oxford University Press, New York.

Knoors H. & Vervloed, M. P. J. (2011) Educational programming for deaf children with multiple disabilities: accommodating special needs. In: *The Oxford Handbook of Deaf Studies, Language and Education* (eds M. Marschark & P. E. Spencer), 2nd edn. **1**, pp. 82–96. Oxford University Press, NY.

Kraijer D. & Bildt A. (2005) The PDD-MRS: an instrument for identification of autism spectrum disorders in persons with mental retardation. *Journal of Autism and Developmental Disorders* **35**, 499–513. doi: 10.1007/s10803-005-5040-0

Landis J. R. & Koch G. G. (1977) The measurement of observer agreement for categorical data. *Biometrics* **33**, 159–74. doi: 10.2307/2529310

Lord C., Rutter M., DiLavore P. C. & Risi S. (1999) *ADOS: Autism Diagnostic Observation Schedule. Manual*. WPS, Los Angeles.

Matson J. L., Dempsey T., LoVullo S. V. & Wilkins J. (2008) The effects of intellectual functioning on the range of core symptoms of autism spectrum disorders. *Research in Developmental Disabilities* **29**, 341–50. doi: http://dx.doi.org/10.1016/j.ridd.2007.06.006

Matson J. L. & Shoemaker M. (2009) Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities* **30**, 1107–14. doi: http://dx.doi.org/10.1016/j.ridd.2009.06.003

Matson J. L. & Williams L. W. (2014) The making of a field: the development of comorbid psychopathology research for persons with intellectual disabilities and autism. *Research in Developmental Disabilities* **35**, 234–8. doi: http://dx.doi.org/10.1016/j.ridd.2013.09.043

Medeiros K., Rojahn J., Moore L. L. & van Ingen D. J. (2014) Functional properties of behaviour problems depending on level of intellectual disability. *Journal of Intellectual Disability Research*, **58**, 151–61. doi: 10.1111/jir.12025

Munde V. & Vlaskamp C. (2014) Initiation of activities and alertness in individuals with profound intellectual and multiple disabilities. *Journal of Intellectual Disability Research, n/a-n/a.* doi: 10.1111/jir.12138

Murdoch H. (1997) Stereotyped behaviours: how should we think about them? *British Journal of Special Education* **24**, 71–5. doi: 10.1111/1467-8527.00018

Neerinckx H., Vos P., Van Den Noortgate W. & Maes, B. (2014) Temporal analysis of attentional processes in spontaneous interactions between people with profound intellectual and multiple disabilities and their support workers. *Journal of Intellectual Disability Research* **58**, 721–33. doi: 10.1111/jir.12067

Nunnally J. C. & Bernstein I. H. (1994) *Psychometric Theory*, 3rd edn. McGraw-Hill, New York.

Pérez-Pereira, M. & Conti-Ramsden, G. (1999) *Language Development and Social Interaction in Blind Children*. Psychology Press, East Sussex, UK.

Peterson C. C. & Siegal M. (1995) Deafness, conversation and theory of mind. *Journal of Child Psychology and Psychiatry* **36**, 459–74. doi: 10.1111/j.1469-7610.1995.tb01303.x

Peterson C. C. & Siegal M. (2000) Insights into theory of mind from deafness and autism. *Mind & Language* **15**, 123–45. doi: 10.1111/1468-0017.00126

Rogers S. J. & Ozonoff S. (2005) Annotation: what do we know about sensory dysfunction in autism? A critical review of the empirical evidence. *Journal of Child Psychology and Psychiatry* **46**, 1255–68. doi: 10.1111/j.1469-7610.2005.01431.x

Roper L., Arnold P. & Monteiro B. (2003) Co-occurrence of autism and deafness. *Autism* **7**, 245–53. doi: 10.1177/1362613030070030002

Rutgers A. H., Bakermans-Kranenburg M. J., van Ijzendoorn M. H. & van Berckelaer-Onnes I. A. (2004) Autism and attachment: a meta-analytic review. *Journal of Child Psychology and Psychiatry* **45**, 1123–34. doi: 10.1111/j.1469-7610.2004.t01-1-00305.x

Stor P. & Storsbergen H. (2006) *Onveilig gehecht of een hechtingsstoornis, het onderkennen van hechtingsproblematiek bij mensen met een verstandelijke beperking*. LEMMA, Utrecht.

Tadić V., Pring L. & Dale, N. (2010) Are language and social communication intact in children with congenital visual impairment at school age? *Journal of Child Psychology and Psychiatry* **51**, 696–705. doi: 10.1111/j.1469-7610.2009.02200.x

Tröster H., Brambring M. & Beelmann A. (1991) The age dependence of stereotyped behaviours in blind infants and preschoolers. *Child: Care, Health and Development* **17**, 137–57. doi: 10.1111/j.1365-2214.1991.tb00684.x

Turner M. (1999) Annotation: repetitive behaviour in autism: a review of psychological research. *Journal of Child Psychology and Psychiatry* **40**, 839–49. doi: 10.1111/1469-7610.00502

Vaccari C. & Marschark M. (1997) Communication between parents and deaf children: implications for social-emotional development. *Journal of Child Psychology and Psychiatry* **38**, 793–801. doi: 10.1111/j.1469-7610.1997.tb01597.x

van Gent T. (2012) Mental health problems in deaf and severely hard of hearing children and adolescents. Findings on prevalence, pathogenesis and clinical complexities, and implications for prevention, diagnosis, and intervention. Doctoral Dissertation, Leiden University, Leiden, Drukkerij Mostert.

Vervloed M. P. J. & Knoors H. (2011) Educational programming for deaf children with multiple disabilities: accommodating special needs. In: *The Oxford Handbook of Deaf Studies, Language and Education* (eds M. Marschark & P. E. Spencer), 2nd edn. **1**, pp. 82–96, Oxford University Press, NY.

Vig S. & Jedrysek E. (1999) Autistic features in young children with significant cognitive impairment: autism or mental retardation? *Journal of Autism and Developmental Disorders* **29**, 235–48. doi: 10.1023/a:1023084106559

Warren D. H. (1994) *Blindness and Children: An Individual Differences Approach*. Cambridge University Press, Cambridge, UK.

Wing L., Gould J., Yeates S. R. & Brierly L. M. (1977) Symbolic play in severely mentally retarded and in autistic children. *Journal of Child Psychology and Psychiatry* **18**, 167–78. doi: 10.1111/j.1469-7610.1977.tb00426.x

Wolters N., Knoors H. E. T., Cillessen A. H. N. & Verhoeven L. (2011) Predicting acceptance and popularity in early adolescence as a function of hearing status, gender, and educational setting. *Research in Developmental Disabilities* **32**, 2553–65. doi: 10.1016/j.ridd.2011.07.003

Zeanah C. H., Smyke A. T., Koga S. F., Carlson E. & The Bucharest Early Intervention Project Core, G. (2005) Attachment in institutionalized and community children in Romania. *Child Development* **76**, 1015–28. doi: 10.1111/j.1467-8624.2005.00894.x

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